



**eResearch: the open access repository of the
research output of Queen Margaret University, Edinburgh**

This is an author-formatted version of an article published as:

McCann, Joanne, Wood, Sara E., Hardcastle, William J., Wishart, Jennifer G. & Timmins, Claire (2008) The relationship between speech, oromotor, language and cognitive abilities in children with Down's syndrome.

Accessed from:

<http://eresearch.qmu.ac.uk/155/>

Repository Use Policy

The full-text may be used and/or reproduced, and given to third parties for personal research or study, educational or not-for-profit purposes providing that:

- The full-text is not changed in any way
- A full bibliographic reference is made
- A hyperlink is given to the original metadata page in eResearch

eResearch policies on access and re-use can be viewed on our Policies page:
<http://eresearch.qmu.ac.uk/policies.html>

**Copyright © and Moral Rights for this article are retained
by the individual authors and/or other copyright owners.**

<http://eresearch.qmu.ac.uk>



Speech Science Research Centre

The relationship between speech, oromotor, language and cognitive abilities in children with Down's syndrome

Joanne McCann, Sara E. Wood,
William J. Hardcastle,
Jennifer G. Wishart & Claire Timmins

Working Paper WP-15

August 2008



Queen Margaret University

EDINBURGH

Update and Sourcing Information August 2008

This paper is available online in pdf format

- 2008 onwards at <http://www.qmu.ac.uk/ssrc>

Author Contact details:

- 2008 onwards at jmccann@qmu.ac.uk
Phone: 0131 474 0000

Subsequent publication & presentation details:

- Details to come.

© the individual authors 2008

This series consists of unpublished “working” papers. They are not final versions and may be superseded by publication in journal or book form, which should be cited in preference.

All rights remain with the author(s) at this stage, and circulation of a work in progress in this series does not prejudice its later publication.

Comments to authors are welcome.

The relationship between speech, oromotor, language and cognitive abilities in children with Down's syndrome

Joanne McCann¹, Sara E. Wood¹, William J. Hardcastle¹,
Jennifer G. Wishart² & Claire Timmins¹

1. Speech Science Research Centre, Queen Margaret University, Edinburgh, UK
2. University of Edinburgh, UK

Abstract

Background: Children and young people with Down's syndrome (DS) present with deficits in expressive speech and language, accompanied by strengths in vocabulary comprehension compared to nonverbal mental age. Speech intelligibility is particularly impaired, but whether speech is delayed or disordered is a controversial topic. Most studies suggest a delay, but no studies explore the relationship between cognitive or language skills and intelligibility. This study sought to determine whether severity of speech disorder correlates with language and cognitive level and to describe the types of errors, developmental or non-developmental, that occur in the speech of children and adolescents with DS.

Methods & Procedures: 15 children and adolescents with DS (aged 10 to 18) were recruited. Participants completed a battery of standardised speech, language and cognitive assessments. The phonology assessment was subject to process analyses. Results from each test were correlated to determine relationships.

Outcome & Results: People with DS present with deficits in receptive and expressive language that is not wholly accounted for by their cognitive delay. Receptive vocabulary is a strength in comparison to language skills, but it was unclear whether it is more advanced compared to non-verbal cognitive skills. The majority of speech errors were developmental in nature but all of the children with DS showed at least one atypical or non-developmental speech error.

Conclusions: Children with DS present with speech disorders characterised by (often unusual) atypical errors alongside many developmental errors. Lack of correlation between speech and cognition or language suggests that the speech disorder in Down's syndrome is not simply due to cognitive delay.

The relationship between speech, oromotor, language and cognitive abilities in children with Down's syndrome

1. Introduction

Down's syndrome (DS) is the most common cause of intellectual impairment, affecting 1 in every 732 live births (Canfield, 2006). It is a genetic disorder, caused by the presence of an extra chromosome in the 21st pair. The degree of cognitive impairment varies widely in people with DS, but 80% present with a moderate intellectual impairment (Roizen, 2002). Individuals with DS present with a specific behavioural phenotype which differs from other syndromes. Theoretically, it is important to know which aspects of the behavioural phenotype are specific to DS in order to learn more about the genetic profile of the syndrome (Abbeduto et al. 2001). Clinically, this is important because knowledge about what areas of functioning are likely to be most or least impaired enables clinicians to design interventions that target areas of weakness and utilise areas of strength in teaching methods. Similarly, it is important to know whether areas of functioning are delayed or disordered. A disordered profile may suggest that spontaneous improvements are less likely and that specific interventions may have to be designed.

Recent research suggests that children and young people with Down's syndrome (DS) present with deficits in expressive speech and language, and strengths in vocabulary comprehension compared to nonverbal mental age (Chapman, 2006). Speech intelligibility is particularly impaired (Rondal and Edwards, 1997) and a survey of families by Kumin (1994) revealed that over 95% of parents reported that their children had difficulty being understood, sometimes or frequently, by people outside of their immediate circle. Whether speech is delayed or disordered is a controversial topic, most studies have suggested a delay (Van Borsel, 1996), or a delay with some elements of disorder (Roberts et al., 2005).

Speech Disorder in DS.

The reduced intelligibility in DS is thought to result from impairments in almost all of the systems required for successful speech. In addition to specific behavioural characteristics, people with DS present with a specific anatomical profile that may affect speech production (Spender et al. 1995, Miller, Leddy and Leavitt, 1999). The ability to create the precise articulations required for speech may be influenced by a smaller than average oral cavity (which gives the impression of a larger tongue), hypotonia of muscles around the mouth, fusion of lip muscles and extra lip musculature. Differences in nerve innervation contribute to reduced speed and range of movement, suggesting that dysarthria may be a factor in reduced intelligibility. Moreover, an increased incidence of hearing impairment in the DS population

(Roizen, 1997) may contribute to the speech and language problems. Although Chapman (1998) estimates that hearing loss accounts for only 4 to 7% of the variance in grammar comprehension, the contribution to intelligibility is less well known.

In addition to the anatomical differences, people with Down's syndrome perform poorly in most areas of motor functioning (Frith & Frith, 1974; Spender et al, 1995; Spano et al, 1999) and particularly motor control in speech production (Kumin, 1994). Barnes et al (2006) found that boys with Down's syndrome showed significantly lower levels of lip, tongue, velopharynx, larynx and coordinated speech function than typically developing boys and lower levels of coordinated speech movements than boys with Fragile X (another common cause of intellectual disability).

A recent survey by Kumin (2006) showed that the majority of children with DS showed signs of dyspraxia (childhood apraxia of speech) but this disorder is rarely diagnosed in DS. Clearly more research is needed to clarify the nature of the speech disorder in DS in order to design appropriate interventions.

Phonological Delay versus Disorder

Differences in anatomy and motor functioning do not in themselves account for the severity of speech disorder often evidenced in DS (Laws and Bishop, 2004). There have been many studies which have suggested that the speech difficulties are a result of a phonological delay (i.e. following the same pattern of development as normal speakers but slower) (e.g. Stoel-Gammon, 1980; Van Borsel, 1996). Others have suggested a phonological delay with some elements of disorder (i.e. following an idiosyncratic developmental pattern, different from normal speakers) (Roberts et al., 2005).

Although it is clear that the speech disorder in DS is not solely the result of a phonological impairment, there has been a great deal of interest in this area. The nature of the phonological errors is controversial. Van Borsel (1996) argues strongly that phonology is delayed in DS. Rather than matching participants with DS (aged 15;4-28;3) to typical children on cognitive measures, his control group consisted of children young enough to still be in the process of phonological acquisition (aged 2;6-3;4). Many errors were similar between the two groups and the phonemes in error were significantly similar. However there was a difference between groups in the frequency of distortions and additional distortions were made by the DS group e.g. 'denasalisation', 'dentalisation' and 'wet'. Despite this, Van Borsel concluded that phonology was delayed in DS. This conclusion is problematic since distortions are usually thought of as phonetic rather than phonological errors, however he acknowledged the uncommon distortions present in the DS speakers may relate to anatomical differences.

Dodd and Thompson (2001) argue convincingly that the speech disorder in DS is not simply a delay but a disorder of phonological acquisition. They compared children with DS to children with inconsistent phonological disorder matched for gender and socio-economic status. As groups, there was no significant difference between percentage consonants correct, confirming that both groups had a similar severity of speech disorder. Both groups of children were inconsistent when producing the same set of words on three different occasions. All of the children with DS were inconsistent, with a mean inconsistency score of 67%. In comparison, a third group of children with a straightforward delay in phonology had inconsistency ratings of less than 20%, suggesting that the inconsistency in DS is not due to delay. Dodd and Thompson suggest that this inconsistency has a different

cause than that seen in children who have inconsistent phonological disorder but are otherwise typically developing. They suggest that underspecified, or “fuzzy” phonological representations may be responsible for the inconsistency or that a difference in language learning environment means that inconsistency is inadvertently reinforced.

The study by Dodd and Thompson presents convincing evidence that the speech disorder in DS is not merely a result of a cognitive disability. If this were the case then we would expect to find no correlation between severity of speech disorder and cognitive level. However, most research does not address the question of whether speech intelligibility is related to language or cognitive level. Anecdotal reports from parents suggest that the most unintelligible children are not necessarily the children with the most severely impaired language or cognitive skills. It was therefore the principal aim of this study to determine whether severity of speech disorder correlates with language and cognitive level and to describe the types of errors, developmental or non-developmental, that occur in the speech of children and adolescents with DS. A second aim was to describe the speech, language and cognitive profiles in children and adolescents with DS to confirm whether the participants in this study conform to the notion that people with DS present with deficits in expressive language and strengths in receptive vocabulary.

2. Method

Participants

Fifteen children with Down’s syndrome (DS) living in the central belt of Scotland participated in the study. The children were aged 9.83-18.75 years (mean 14.3, SD 3.07), and the group comprised of 12 boys and 3 girls. Children were excluded if any of the following criteria applied: (1) English was not the child’s first language and the main language of the home; (2) there was evidence of severe hearing loss (aided threshold >40 dB); (3) was not able to use single words (i.e. no speech); (4) there was a co-morbid diagnosis of autism. Most children had undergone recent audiological testing which confirmed their hearing status. However, to confirm adequate speech perception ability, all of the children completed the Manchester Picture Test (Hickson, 1987).

As part of a larger study investigating speech motor control all of the children had custom-made electropalatography palates (EPG palates). EPG records the timing and location of tongue with the hard palate. All of the children were wearing their EPG palates during the recordings of the phonology test (see below).

Standardised Assessments

Language, speech and cognitive assessments

All children completed a battery of standardised speech, language and cognitive assessments. Speech and language tests were carried out by a qualified Speech and Language Therapist (the first author); cognitive assessments were carried out by a child psychologist. Most children completed the battery in three, one hour sessions allowing for breaks as requested by either the child or their carer. In order to accommodate the severe language and cognitive impairment typical in DS, in most cases the assessments used were standardised on much younger children. Age equivalent scores, raw scores and percentages were therefore used for the analyses.

Cognitive Ability

Cognitive ability was assessed using the full form of the Weschler Preschool and Primary Scale of Intelligence (WPPSI-IIIUK, Weschler, 2003). Verbal, performance and full-scale age equivalents were calculated.

Receptive Vocabulary

The British Picture Vocabulary Scales-II (BPVS-II, Dunn et al., 1997) were used as a measure of receptive vocabulary. This assessment covers a wide age range and is a well-established tool for measuring verbal mental age. It is a multiple-choice test in which participants must select one of four pictures to match a single word spoken by the tester.

Receptive and Expressive Language

The Clinical Evaluation of Language Fundamentals-Preschool UK (CELF-P, Wiig, Secord & Semel, 1992) was used to measure receptive and expressive language. This test allows calculation of receptive, expressive and general language age equivalents.

Phonology

All children completed the phonology subtest of the Diagnostic Evaluation of Articulation and Phonology (DEAP, Dodd, Hua, Crosbie, Holm and Ozanne, 2002). This is a measure of consonant production in single words, covering most consonants of English in word initial and final positions. The phonology subtest allows calculation of percentage consonants correct (PCC); percentage vowels correct (PVC); percentage phonemes correct (PPC) and single words/ connected speech phoneme agreement (SvC). Audio recordings were made to allow for fine phonetic transcription. All of the children were wearing EPG palates during the completion of the DEAP, however they had undergone a programme of acclimatisation to the palate prior to the recording.

Error Analysis

All errors produced in the phonology subtest of the DEAP were subjected to a process analysis and classified as either typical (occurring in the speech of children aged 2;0 to 5;11) or atypical (occurring in less than 10% of typical children aged 2;0 to 5;11) using data from Dodd et al. (2002). Although all of the children's errors were described in terms of process analyses this does not necessarily suggest that the errors are a result of a phonological impairment. While some errors were thought to be phonological in nature, for example fronting of /k/ to [t], other processes were more likely to be phonetic in nature, for example lateralisation of sibilants. For the purposes of the analysis all errors were counted together. In addition to calculating the number of times a process occurred, the number of children displaying a process 3 or more times (Dodd et al. 2002) was also calculated. This enabled us to identify whether errors occurred only occasionally in a child's speech or whether they were more prevalent. It also allowed us to determine whether particular processes were common to all or most children with DS.

Oromotor Function

Oromotor function was assessed using the Clinical assessment of oropharyngeal motor development in young children (Robbins & Klee, 1987). In this assessment children are required to perform speech and non-speech oral movements which are scored as either adult-like (2 points), approaching adult-like (1 point) or absent (0). Raw scores were converted to a percentage.

Intelligibility

Previous studies of speech in DS have used parent questionnaires to rate intelligibility (Kumin 1994). We sought to use a standardised method in order to quantify the severity of the unintelligibility, and to enable us to compare percentage consonant correct (from the DEAP) with more global intelligibility. Since many of the young people with DS spoke in either single words or short phrases, the Children's Speech Intelligibility Measure (CSIM, Wilcox & Morris, 1999) was chosen. The test involves a listener who is unfamiliar with the child listening to 50 (imitated) words and identifying which word was uttered from a possible 12 phonetically-similar words (for each of the 50 words). Percentage of correctly identified words was calculated.

3. Results

Table one shows the group results for all measures, with numbers expressed as age equivalents or percentages as appropriate. Most of the children failed to meet the basal age equivalent on the DEAP (3 years), meaning that mean age equivalents (AE) could not be calculated for this test. As the CSIM is not standardised on typical children no age equivalents were available for this test. As can be seen from the table, there was a wide range in ability in the cognitive test, with full scale cognitive ability ranging from 2.58 years AE to 7.17 years. A similar range was found for the receptive vocabulary measure (BPVS) ranging from 2.83 years to 7.2 years. However the highest score achieved on the CELF Receptive Language measure was only 4.83 years AE. None of the language or cognitive measures correlated with chronological age, suggesting that language and cognitive skills are fairly stable in the 9-18 years age group.

Table One: Standardised Assessment Results.

		DEAP						CELF			WPPSI		
	CHRON AGE	PCC	PVC	PPC	SvC	CSIM	BPVS	CELFE	CELFC	RK	VIQ	PIQ	FSIQ
Age Eq Range Mean (SD)	9.83-18.75 14.28 (3.07)	<3-4.0	<3-4.0	<3-4.0	<3-3.5		2.83-7.25 4.78 (1.63)	2.5-4.42 3.35 (0.63)	2.75-4.83 3.82 (0.69)		2.58-7.17 4.53 (1.53)	2.58-7.17 5.43 (1.34)	2.58-7.17 4.96 (1.10)
% Range % Mean (SD)		12.93-87.92 53.83 (24.13)	47.30- 100 84.12 (16.67)	26.67-92.12 64.04 (20.41)	0-80 43.64 (27.20)	20.0-84.09 52.59 (23.66)	13.10-44.05 28.25 (10.90)	13.21-67.92 42.79 (17.11)	32.26- 87.10 64.75 (20.27)	51.92-91.35 71.99 (12.55)			

DEAP=Diagnostic Evaluation of Articulation and Phonology; PCC= Percentage Consonants Correct; PVC= Percentage Vowels Correct; PPC=Percentage Phonemes Correct; SvC= Single Word/ Connected speech agreement.

CSIM= Children's Speech Intelligibility Measure.

BPVS= British Picture Vocabulary Scale-II

CELF= Clinical Evaluation of Language Fundamentals-Preschool UK; CELF-E= CELF Expressive Language; CELFC= CELF Receptive Language

RK= Robbins & Klee Clinical assessment of oropharyngeal motor development in young children.

WPPSI= Wechsler Preschool and Primary Scale of Intelligence; VIQ=Verbal Intelligence; PIQ= Performance Intelligence; FSIQ= Full-Scale Intelligence

Pearson's correlations were used to test for significant correlations between all the measures. A threshold of $p < 0.01$ was taken as significant unless otherwise stated. Tables two and three show significant correlations between measures.

Table Two: Correlations: Language and Cognitive Measures.

	BPVS	CELF	CELF Exp	CELF Rec	VIQ
PIQ					
VIQ	$r = .697; p = .006$				
CELF Rec	$r = .826; p < .0005$	$r = .887; p < .0005$	$r = .786; p = .0001$		
CELF Exp	$r = .885; p < .0005$	$r = .967; p < .0005$			
CELF	$r = .875; p < .0005$				

PIQ= Performance IQ

VIQ= Verbal IQ

CELF Rec= Clinical Evaluation of Language Fundamentals- receptive language

CELF Exp= Clinical Evaluation of Language Fundamentals- expressive language

CELF = Clinical Evaluation of Language Fundamentals- total language

Table Three: Correlations: Speech and oromotor measures

	PCC	PPC	PVC	SvC	RK
CSIM	$r = .889; p < .0005$	$r = .876; p < .0005$	$r = .694; p = .012$	$r = .733; p = .007$	$r = .945; p < .005$
RK	$r = .826; p < .0005$	$r = .801; p < .0005$	$r = .620; p = .014$	$r = .720; p = .002$	
SvC	$r = .831; p < .0005$	$r = .811; p < .0005$	$r = .672; p = .002$		
PPC	$r = .985; p < .0005$				

PCC= Percentage Consonants Correct

PPC= Percentage Phonemes Correct

PVC= Percentage Vowels Correct

SvC= Single words/connected speech agreement

RK= Robbins Klee, oromotor assessment

CSIM= Children's Speech Intelligibility Measure

Language and cognitive measures

The language measures correlated highly with each other (BPVS-II and CELF [$r = 0.875; p < .0005$]). Within the CELF, expressive language correlated highly with receptive language (CELF expressive and CELF receptive [$r = .786; p = .001$]). The BPVS-II receptive vocabulary measure correlated highly with verbal IQ composite of the WPSSI-III (BPVS-II and WPSSI-III VIQ [$r = .697; p = .006$]) but the CELF did not correlate with

VIQ ($r=.500$; $p=.082$). WPPSI Performance IQ (PIQ) did not correlate with any of the language measures. This suggests that receptive and expressive language are related but non-verbal ability is independent of other skills.

Paired t-tests were used to determine, in terms of age equivalents, which language and cognitive measures showed relatively greater levels of impairment. A Bonferroni correction was applied, adjusting the significance level to $p<0.003$. Performance IQ was greater than verbal IQ in 9 out of the 15 children and equivalent in two of the children. Despite this, there was no significant difference between PIQ and VIQ ($p=.066$) but non-verbal cognitive skills were in advance of language skills (PIQ and CELF total: ($p<.0005$). An exception to this was receptive vocabulary skills as measured by the BPVS which were commensurate with cognitive skills (VIQ [$p=.451$]; PIQ [$p=.125$] and FSIQ [$p=.524$]).

BPVS receptive vocabulary was significantly in advance of expressive and receptive language (CELF receptive [$p<.0005$] and CELF expressive [$p=.002$] respectively) and receptive language as measured by the CELF was in advance of expressive language ($p=.001$).

Speech measures

As two thirds (10) of the children with DS failed to meet the basal age equivalent of 3;0 years in the DEAP percentage consonants correct, age equivalents are not reported for this measure. This suggests that the majority of the children with DS presented with very severe speech disorders. In order to determine whether a relationship existed between cognitive, language and speech skills the measures from the DEAP and CSIM were correlated with the language and cognitive measures. Since floor age equivalent results were obtained in the DEAP, raw or percentage scores were used for the calculations.

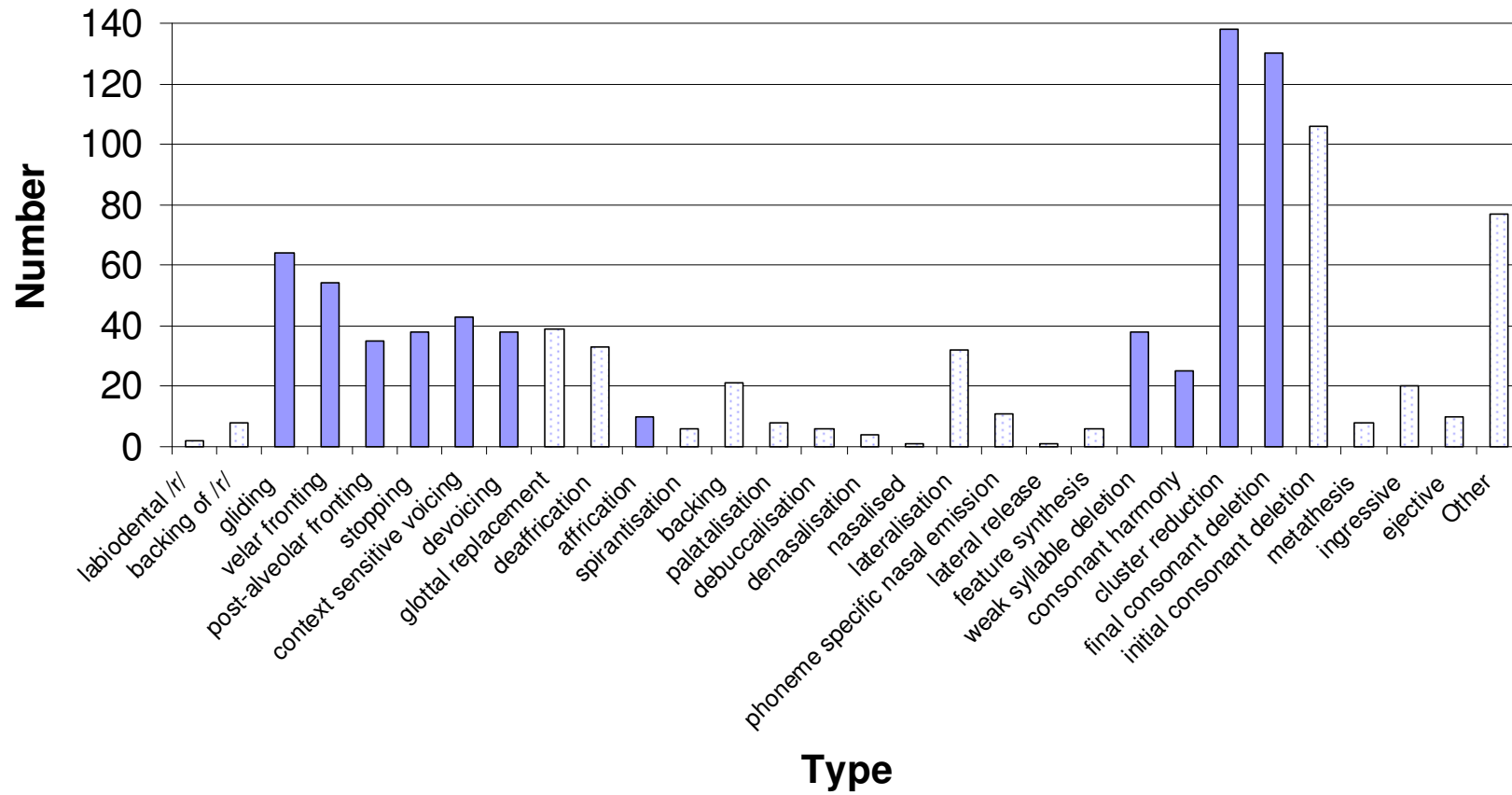
All of the measures from the DEAP, PCC, PVC, PPC and SvC, correlated highly with each other (all $p<.0005$). The oromotor measure, the Robbins Klee, correlated highly with PCC, PPC and SvC ($r=.827$; $p<.0005$; $r=.801$; $p<.0005$ and $r=.720$; $p=.002$ respectively) and weakly with PVC ($r=.620$; $p=0.14$). Results from the CSIM correlated highly with all of the measures from the DEAP (PCC [$r=.889$; $p<.0005$]; PVC [$r=.694$; $p=.012$]; PPC [$r=.867$; $p<.0005$]; SvC [$r=.733$; $p=.007$]) and with the Robbins Klee ($r=.945$; $p<.005$) This suggests that children with poorer oromotor skills (speech and non-speech) produced less intelligible speech with more errors.

PCC did not correlate with any of the language measures (BPVS [$r=.377$; $p=.166$], CELF exp [$r=.299$; $p=.299$], CELF rec [$r=.364$, $p=.200$]) nor did the CSIM (BPVS [$r=.500$; $p=.098$], CELF exp [$r=.512$; $p=.107$], CELF rec [$r=.577$, $p=.063$]) suggesting that speech disorder is independent of language ability. Moreover PCC did not correlate with performance or full-scale IQ ($[r=.035$; $p=.901$] and $[r=.330$; $p=.229$] respectively) although there was a weak correlation with verbal IQ ($r=.576$; $p=.025$). Moreover, there was no correlation between the CSIM and performance or full-scale IQ ($[r=.182$; $p=.571$] and $[r=.378$; $p=.225$] respectively) although again there was a weak correlation with verbal IQ ($r=.579$; $p=.049$).

Error Analysis.

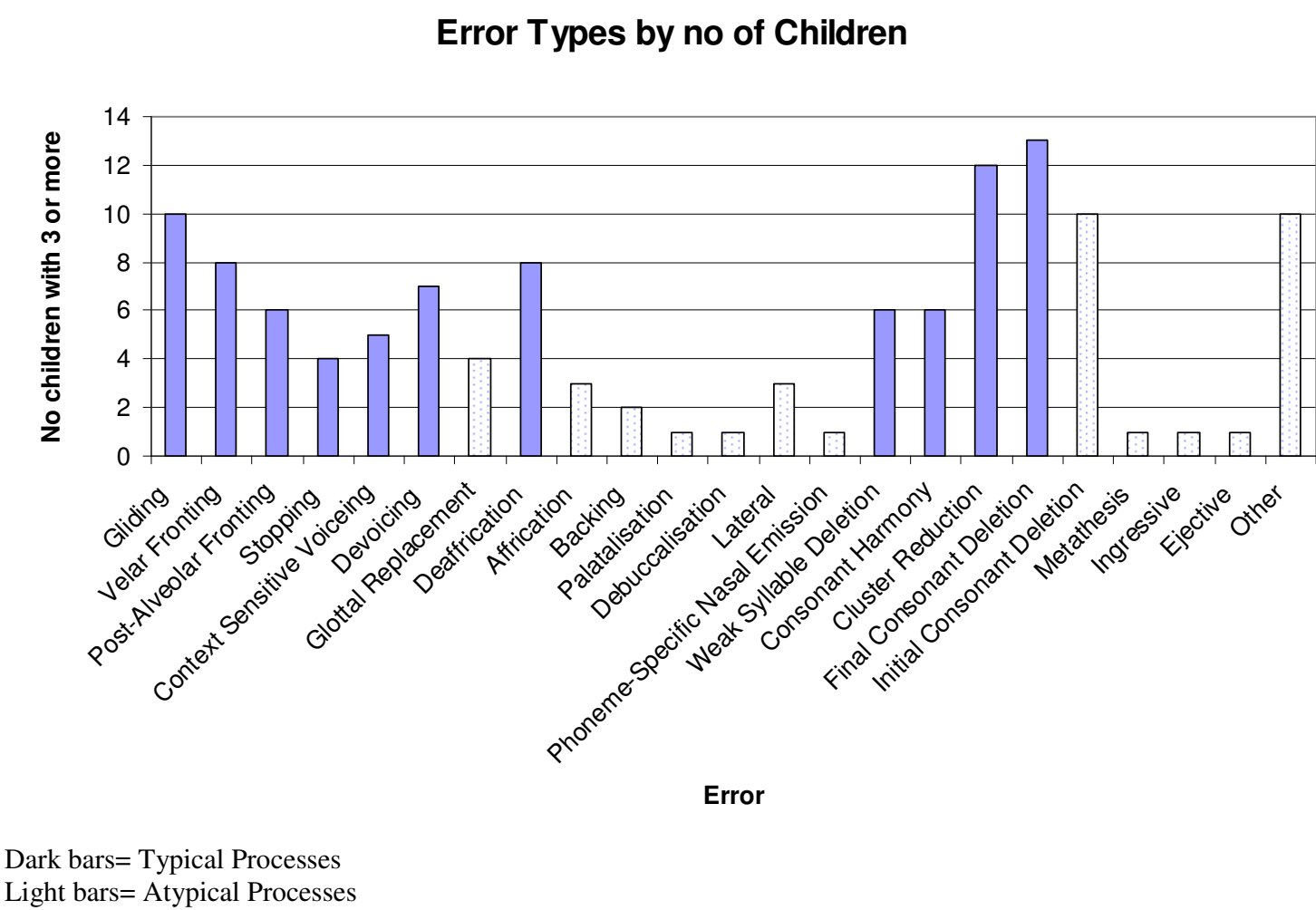
Twenty-nine different processes were identified in the single word productions of the DEAP phonology subtest. A further 65 speech errors (7.61% of total errors) were unclassifiable due to their unusual nature. Of these 29 different processes, 23 were evident at least three times in one or more child's speech. Figure One shows the frequency of the different phonological processes and Figure Two shows the number of children producing each process at least three times.

Figure One: Phonological Processes



Dark bars= Typical Processes
 Light bars= Atypical Processes

Figure Two.



Cluster reduction was the most common process (13.64% of the errors, 12 children), followed by the other structural simplification processes: final consonant deletion (12.85%, 13 children), initial consonant deletion (10.47%, 10 children) and then gliding (6.32%, 10 children). Despite this there was no significant difference between the number of systemic and structural errors ($p=.057$). The majority of processes (66.23%, paired samples t-test, $p=.021$) were those commonly found in younger typically developing children (Dodd et al. 2002) suggesting mainly a delayed pattern of development, however all of the children also presented with atypical or non-developmental errors. Only one child had more atypical (75%) than typical errors. This particular child's speech was characterised by deletion of word initial fricatives, production of other fricatives as ingressive, and production of word-final stops as ejectives. There was no correlation between the number of typical errors and the number of atypical errors ($r=.330$; $p=.230$) suggesting that contrary to what might be expected children with more developmental errors did not also present with more atypical errors.

4. Discussion

The children with DS presented with widely varying ability. Full scale cognitive ability was in the range of 2.58 years to 7.17 confirming results of earlier studies (Chapman and Hesketh, 2001). Results of the language and cognitive tests broadly support the literature which suggests that children and young people with Down's syndrome (DS) present with deficits in expressive language (Chapman, 2006). Expressive language was impaired not only in relation to non-verbal cognitive ability but also in relation to receptive language. Since many of the children were highly unintelligible it is difficult to know if this represents a real discrepancy in expressive language or whether the children are simply saying less as a strategy to make themselves more easily understood. In contrast to previous research, we did not find a relative strength in receptive vocabulary compared to non-verbal ability but we did find that receptive vocabulary was superior to expressive and receptive language. As predicted, language skills correlated highly but, rather unexpectedly, language skills did not correlate with performance IQ. Theoretically this is important because it suggests that the language impairment in DS is not simply a result of cognitive delay but some other factor, essentially a 'specific' language impairment. Clinically this is important because it suggests that language intervention may be warranted in people with DS who present with a discrepancy between language and cognition.

Speech

Severe speech disorders were evident in the participants. Most of the children did not meet the basal age equivalent of the test used (3 years). This makes it difficult to determine statistically whether speech is more impaired than language or cognitive skills but this seems likely to be the case given that most of the children with DS performed above an age equivalent of 3 years in the language and cognitive assessments. Furthermore, severity of speech disorder (both the DEAP and the CSIM) did not correlate with any of the language or cognitive measure, suggesting that the speech impairment is caused by some factor other than language or cognitive delay. One possible factor may

be reduced oromotor skills, confirmed by correlations between the measure of oromotor function, percentage consonants correct and the speech intelligibility measure.

The intelligibility measure correlated highly with percentage consonants correct. This suggests that children who perform poorly in the phonology test are also less intelligible to an unfamiliar listener. It also gives confidence that single word phonology tests, which are often used to diagnose speech disorders, are reflective of more general intelligibility which is often not measured in the speech and language therapy clinic.

Process analyses.

Twenty-nine different processes were identified in the speech of the children with DS, with 23 of these occurring at least 3 times in at least one child's speech. Of these only eleven (37.93%) were processes found in typically developing children. A further 65 errors were not able to be classified as a known process (Grunwell, 1985) because of their unusual or idiosyncratic nature.

Structural processes (consonant deletions) were very common, including the non-developmental process of initial consonant deletion. In the most severely affected children words were produced without any consonants at all, preserving only the vowel in the case of a CVC structure.

There was a greater incidence (number of errors) of the processes usually found in typical development and all of the children bar one had more developmental than non-developmental errors. This could be interpreted as a case of delay rather than disorder (Van Borsel, 1996), however, given that all of the children with DS had at least one atypical error it seems premature to conclude this. Moreover, the severity and pervasiveness of the delayed error patterns may in itself be enough to suggest that speech in DS is severely disordered. When a child presents with a single disordered speech error in the face of many delayed patterns this is usually diagnosed by speech and language therapists as a speech disorder. For example, four of the children with DS presented with phoneme specific nasal emission, a rare functional articulation disorder where air is emitted through the nose rather than the mouth when saying specific sounds. Since the nasal emission is confined to certain phonemes (usually sibilants) the cause cannot be organic, also, since there is no loss of contrast this error in itself may not affect intelligibility. However, the resultant speech is so unusual sounding that it may impact on social acceptance. Dodd and Thompson (2001) suggest that inconsistency in the speech of children with DS may result from incomplete phonological representations, it seems possible that errors like phoneme specific nasal emission may also be the result of underspecified phonological representations. This would be evidence for disorder rather than delay in DS.

Less investigated is the possibility of dyspraxia as a diagnosis in DS. Although the present study was not specifically designed to investigate this, there is some evidence that at least some of the children with DS present with symptoms usually found in dyspraxia (Kumin, 2006). For example, processes that are hard to classify have been suggested to be one of the distinguishing features of dyspraxia (ASHA, 2007) and there were many examples of this in the data. Moreover, most of the children omitted sounds and syllables (Rupela and Manjula, 2007) and many had a limited repertoire of phonemes. In some cases the repertoire was so severely reduced that many words consisted of vowels only. Performance in the oromotor assessment (Robbins and Klee,

1987) was also disordered and most children had difficulty combining and sequencing phonemes in the diadochokinetics tasks in the assessment (maximum performance rate of syllables with altering places of articulation, i.e. p t k p t k).

5. Conclusions

Children and adolescents with Down's syndrome present with deficits in receptive and expressive language that is not wholly accounted for by their cognitive delay. While receptive vocabulary is a strength in comparison to language skills it is unclear whether it is more advanced compared to non-verbal cognitive skills.

Speech is particularly impaired in DS. The finding that all the children with DS show at least one atypical or non-developmental speech error leads us to believe that children with DS present with speech disorders characterised by (often unusual) atypical errors alongside many developmental errors. The cause of the speech disorder in DS remains unclear. However, anecdotal reports that the more unintelligible children are not necessarily the most cognitively or linguistically impaired was confirmed by the lack of a correlation between speech and cognition or language. This suggests that the cause of the speech disorder is not merely a cognitive delay. It is probable that the disorder in DS is multi-factorial or differs in different individuals. Underspecified phonological representations may be responsible in some children, whereas others seem to have difficulty with the motor control required for speech which may warrant a diagnosis of dyspraxia.

From a clinical perspective it seems clear that the speech disorder in DS warrants intervention. Clinicians should apply their skills in the differential diagnosis of speech disorders to children with DS, allowing interventions to target the cause of the disorder in each individual.

Acknowledgements

This research was supported by a grant from the Medical Research Council (G0401388). Thanks to Ann Robertson for administering the cognitive assessments; Down's Syndrome Scotland for their help recruiting participants; and the children and their parents, without whom the research would not have been possible.

References

- ABBEDUTO, L., PAVETTO, M., KESIN, E., WEISSMAN, M., KARADOTTIR, S., O'BRIEN, A. and CAWTHON, S., 2001, The linguistic and cognitive profile of Down syndrome: Evidence from a comparison with fragile X syndrome. *Down Syndrome Research and Practice*, **7**: 9-15.
- AMERICAN SPEECH-LANGUAGE-HEARING ASSOCIATION, 2007, *Childhood Apraxia of Speech* [Technical Report]. Available from www.asha.org/policy.
- BARNES, E., ROBERTS, J., MIRRETT, P., SIDERIS, J. AND MISENHEIMER, J., 2006, A Comparison of Oral Structure and Oral-Motor Function in Young Males with Fragile X Syndrome and Down Syndrome. *Journal of Speech, Language and Hearing Research* **49**:903-917.
- CANFIELD MA, HONEIN MA, YUSKIV N, XING J, MAI CT, COLLINS JS, DEVINE O, PETRINI J, RAMADHANI TA, HOBBS CA, KIRBY RS., 2006, National estimates and race/ethnic-specific variation of selected birth defects in the United States, 1999-2001. *Birth Defects Res A Clin Mol Teratol*, **76**:747-56.
- CHAPMAN, R., 2006, Language learning in Down syndrome: The speech and language profile compared to adolescents with cognitive impairment of unknown origin. *Down Syndrome Research and Practice*, **10**: 61-66.
- CHAPMAN, R. and HESKETH, L., 2001, Language, cognition, and short-term memory in individuals with Down syndrome. *Down Syndrome Research and Practice*, **7**, 1-7
- DODD, B., HUA, Z., CROSBIE, S., and HOLM, A., 2002, *Diagnostic Evaluation of Articulation and Phonology*. London: The Psychological Corporation.
- DODD, B. and THOMPSON, L., 2001, Speech disorder in Down's syndrome. *Journal of Intellectual Disability Research*, **45**: 308-316.
- DUNN, L., DUNN, L., WHETTON, C., and BURLEY, J., 1997, *British picture vocabulary scale – second edition*. Windsor: NFER-Nelson.
- FRITH U AND FRITH CD, 1974, Specific Motor disabilities in Down's Syndrome. *Journal of Child Psychology and Psychiatry* **15**:293-301.
- GRUNWELL, P., 1985, *Phonological Assessment of Child Speech (PACS)*. Windsor: NFER-Nelson
- HICKSON, F., 1987, *The Manchester Picture Test: A Summary*. Manchester: University of Manchester.
- KUMIN, L., 1994, Intelligibility of speech in children with Down syndrome in natural settings: parents' perspective. *Perceptual Motor Skills*, **78**, 307-13
- KUMIN, L., 2006, Speech intelligibility and childhood verbal apraxia in children with Down syndrome. *Down Syndrome Research and Practice*, **10**, 10-22.
- LAWS, G. and BISHOP, D., 2004, Verbal deficits in Down's syndrome and specific language impairment: a comparison. *International Journal of Language and Communication Disorders*, **39**, 423-451.
- MILLER, J., LEDDY, M. and LEAVITT, L., 1999, *Improving the communication of people with Down syndrome*. Baltimore: Paul H. Brookes Pub.
- ROBERTS, J., LONG, S., MALKIN, C., BARNES, E., SKINNER, M., HENNON, E. AND ANDERSON, K., 2005, A Comparison of Phonological Skills of Boys with Fragile X Syndrome and Down Syndrome. *Journal of Speech, Language, and Hearing Research*, **48**, 980-995.

- ROBBINS, J. and KLEE, T., 1987, *Clinical assessment of oropharyngeal motor development in young children*. *Journal of Speech and Hearing Disorders*, **52**, 271-277.
- RONDAL, J. and EDWARDS, S., 1997, *Language in Mental Retardation*. Whurr: London.
- ROIZEN, N., 1997, Hearing loss in children with Down syndrome: a review. *Down Syndrome Quarterly*, **2**, 1-4.
- ROIZEN, N., 2002, Down Syndrome. In: Batshaw, M. (Ed) *Children with disabilities*, 5th Ed. Baltimore, MD: Brookes, 361-376.
- RUPELA, V. and MANJULA, R., 2007, Phonotactic patterns in the speech of children with Down syndrome. *Clinical Linguistics and Phonetics*, **21**: 605-622
- SPANIO, M., MERCURI, E., RANDO, T., PANTO, T., GAGLIANO, A., HENDERSON, S. and GUZZETTA, F., 1999, Motor and Perceptual-motor competence in children with Down syndrome: variation in performance with age. *European Journal of Paediatric Neurology*, **3**: 7-13.
- SPENDER, Q., DENNIS, J., STEIN, A., CAVE, D., PERCY, E. and REILLY, S., 1995, Impaired oral-motor function in children with Down's syndrome: a study of three twin pairs. *European Journal of Disorders of Communication*, **30**, 87-97.
- STOEL-GAMMON C., 1980, Phonological analysis of four Down's syndrome children. *Applied Psycholinguistics* **1**: 31-48.
- VAN BORSEL, J., 1996, Articulation in Down's syndrome adolescents and adults. *European Journal of Disorders of Communication*, **31**, 414-444.
- WESCHLER, D., 2003, *Wechsler Primary and Preschool Scale of Intelligence- Third UK Edition*. London: The Psychological Corporation.
- WIGG, E., SECORD, W. and SEMEL, E., 1992, *Clinical Evaluation of Language Fundamentals- Preschool UK*. London: The Psychological Corporation.
- WILCOX, K., & MORRIS, S., 1999, *Children's Speech Intelligibility Measure (CSIM)*. London: The Psychological Corporation.